

## PEER REVIEW HISTORY

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### ARTICLE DETAILS

<b>TITLE (PROVISIONAL)</b>	Reaching Consensus on Reporting Patient and Public Involvement (PPI) in Research: Methods and lessons learned from the development of reporting guidelines
<b>AUTHORS</b>	Brett, Jo; Staniszewska, Sophie; Simera, Iveta; Seers, Kate; Mockford, Carole; Goodlad, Susan; Altman, Doug; Moher, David; Barber, Rosemary; Denegri, Simon; Entwistle, Andrew; Littlejohns, Peter; Morris, Christopher; Suleman, Rashida; Thomas, Victoria; Tysall, Colin

### VERSION 1 - REVIEW

<b>REVIEWER</b>	Marit By Rise Department of mental health, Norwegian University of Science and Technology, Norway.
<b>REVIEW RETURNED</b>	22-Apr-2017

<b>GENERAL COMMENTS</b>	<p>This is a methods manuscript describing the (further) development of a guideline for how papers should report patient and public involvement (PPI) in research.</p> <p><b>Overall impression:</b> This is a well-written manuscript on an important topic. Since PPI in research projects is increasingly advocated for and sought after, the need for high quality reporting guidelines is evident. This manuscript seeks to answer to this need. While this manuscript has many qualities, I think that a revision would improve important aspects of the manuscript. My comments are as follows:</p> <p><b>Major issues:</b> <b>Patient and public involvement in research</b> 1. Throughout the manuscript it is not specified that the topic is PPI in research and not in general (as in health services etc.). The title is confusing since it states that the manuscript is about guidelines for reporting PPI, and does not specify that the subject is PPI in research in particular. I would recommend that the title is revised to specify this. The sentence "Patient and Public Involvement in health and social care research" could also be interpreted to include all research on PPI, not only when patients or representatives from the public take part in research processes. This makes the Lay summary and the Abstract somewhat unclear, and this lack of specification is present throughout the manuscript.</p> <p><b>Background:</b> 2. The description of the development process is well-written and in general easy to follow. It is clear how the systematic literature review, the Delphi process</p>
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	<p>and the consensus meeting were conducted. However, I miss a description of the transition from the literature review to the Delphi process. The description “jumps” from the review directly to the GRIPP2 guideline items and the first stage of the Delphi process. How were the results from the updated literature review developed into the items in the first Delphi survey, and how did the researchers further develop the responses in the first survey into the second etc.? These methodological steps are important and are now lacking. (In the present manuscript, it is only described that “Two researchers analyzed the results of round 1 and free text comments were examined to inform any potential additional items.” (Page 12, paragraph 2))</p> <p><b>Discussion and conclusion:</b></p> <p>3. The first and second paragraph in this section is only repetition of the argument for doing the study, and should be omitted, except for the last two sentences, which sum up the aim of the paper.</p> <p>4. Too much of the description in the Methods section is repeated in the part “Important aspects and lessons learned from the development process”. The repetition in this section should be removed and only the parts concerning aspects of lessons learned should be kept. This part of the manuscript should in general be shortened to avoid unnecessary repetition.</p> <p>5. On page 20, last paragraph, results from the pilot study are referred to. The pilot study is described at page 11, second paragraph, but it is not described how the results from the pilot were used in the Delphi survey. I suggest that the information about the pilot study in the Discussion section is moved to the Methods section, since it is barely descriptions and not an actual discussion.</p> <p>6. From reading the list of points in “Strengths and limitations of this study” I anticipated more discussion of the role of the facilitator of the consensus meeting. This part of the discussion is short and very general, and includes no information on this specific meeting for this specific project. An addition here could increase the usefulness of this manuscript.</p> <p><b>Minor issues:</b></p> <p>1. INVOLVE is introduced in the Background section without any references. This should be added.</p> <p>2. On page 17, last paragraph is says “Evidence shows only an 8 % improvement in identification of relevant papers when all abstracts are reviewed by two researchers.” It is unclear what the reference for this statement is. This should be clarified.</p>
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<b>REVIEWER</b>	Pam Carter University of Leicester, UK
<b>REVIEW RETURNED</b>	14-May-2017

<b>GENERAL COMMENTS</b>	<p>BMJ Review</p> <p>The objective of this article is to describe methods used in the</p>
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	<p>development of guidance (GRIPP2), to reflect on challenges encountered and to describe lessons learned from the process.</p> <p>The abstract summarises the main article and a lay summary is also provided. The language is in the main very clear and accessible.</p> <p>Exceptions:</p> <p>p.7 line 53 data base is usually expressed as one word, database.</p> <p>p.8 lines 21-23 is missing a word – WERE reviewed?</p> <p>p.9 line 26 educational should be education.</p> <p>p.14 line 36 or 37 – this method IS intended to ...</p> <p>It is difficult to review this article without access to the submitted article that reports the main study – cited as reference number 9 Staniszevska et al. submitted but not yet published. I presume the authors submitted separate articles for reasons of word count limitations but there may be a risk of duplication. For example, a lot of content is devoted to a very clear exposition of the Delphi consensus method and I imagine that this is also covered in the main article.</p> <p>There is a welcome aspect of reflexivity where the authors acknowledge the limitations of their study. Conversely, although PPI is reported here, it is not clear how PPI members of the steering group added to or changed the research.</p> <p>The following areas could be considered:</p> <p>It would be helpful if the article distinguished explicitly between studies where PPI was the main focus of the research and other articles that report PPI as part of research that is aimed at answering a clinical or health services or social services research question.</p> <p>While the article acknowledges the lack of consistent MESH terms, there is no acknowledgement of the way in which different ideologies / discourses or traditions impact on PPI. For example we might expect to see Ives et al cited as well as more recent publications that</p>
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	<p>deal with critical aspects of PPI.</p> <p>Ives, Jonathan, Sarah Damery, and Sabi Redwod. "PPI, paradoxes and Plato: who's sailing the ship?." <i>Journal of medical ethics</i> (2012): medethics-2011.</p> <p>Similarly, Forbat, Liz, Gill Hubbard, and Nora Kearney. "Patient and public involvement: models and muddles." <i>Journal of clinical nursing</i> 18.18 (2009): 2547-2554.</p> <p>Again, it may be that these conceptual issues are covered in the other article that has been submitted but I cannot tell.</p> <p>PPI has long been a contested area and the impulse towards consensus is understandable but readers might learn more than is presented here currently from the challenges faced in resolving difference. This is glossed over in the description of the consensus face to face meeting but perhaps it is discussed in the main article. Materials were sent in advance of the consensus meeting but the authors should specify which the 'one or two key papers' were and reflect on whether and if so, how, these papers were thought to have influenced the discussion.</p> <p>Could the authors explain the need for GRIPP 2 and how frequently this might need to be updated?</p> <p>A weblink to HTAi Citizen and Patient Involvement Group would be helpful – perhaps as a footnote.</p> <p>In the Delphi survey sub section of Methods, Round 3 changed concepts. It would be interesting to explain and discuss these concepts.</p> <p>Finally, as a researcher with an interest in public policy, I would simply urge caution in assuming that policy is necessarily based solely on 'rational decision making' (p.7), value judgements remain important.</p>
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## VERSION 1 – AUTHOR RESPONSE

Reviewer 1

R: The objective of this article is to describe methods used in the development of guidance (GRIPP2), to reflect on challenges encountered and to describe lessons learned from the process.

The abstract summarises the main article and a lay summary is also provided. The language is in the

main very clear and accessible. Exceptions:

p.7 line 53 data base is usually expressed as one word, database.

p.8 lines 21-23 is missing a word – WERE reviewed?

p.9 line 26 educational should be education.

p.14 line 36 or 37 – this method IS intended to ...

A: Thank-you. We have now corrected the mistakes highlighted

R: It is difficult to review this article without access to the submitted article that reports the main study – cited as reference number 9 Staniszewska et al. submitted but not yet published. I presume the authors submitted separate articles for reasons of word count limitations but there may be a risk of duplication. For example, a lot of content is devoted to a very clear exposition of the Delphi consensus method and I imagine that this is also covered in the main article.

A: The results paper, which is being published in the BMJ, is shortened to approximately 2000 words+ in line with other equator guideline papers. There is minimal description of the methods used, and the paper refers to this paper for readers to get more detailed information on methods.

R: There is a welcome aspect of reflexivity where the authors acknowledge the limitations of their study. Conversely, although PPI is reported here, it is not clear how PPI members of the steering group added to or changed the research.

A: This is a good point. This has been explained in the results paper, but we have added in the following paragraph to report how PPI members added or changed the research. The section 'Patient Partners Contribution' has been added at the end of the discussion.

The PPI members contributed to the development of GRIPP in a number of ways. Throughout the initial stages of collating the evidence and identifying items for the GRIPP checklist, the patient partners highlighted the importance of including items referring to the context and processes of PPI, suggesting that this affected the impact that PPI had on research. The patient partners, along with other patient organisations and charities, recruited nearly half of all participants for the Delphi survey. The patient partners helped other patients with the technical aspects of completing the online survey, improving the response rate in each Delphi survey round. The patient partners not only checked the comprehension of the changed items and comments from the lay perspective between rounds, but were also integral to helping the researchers keep to the scheduled time of the Delphi survey. Throughout the write up phase for both the results paper and the methods paper, the patient partners contributed to the lay sections and contributed to edits of the paper.

R: The following areas could be considered:

It would be helpful if the article distinguished explicitly between studies where PPI was the main focus of the research and other articles that report PPI as part of research that is aimed at answering a clinical or health services or social services research question.

A: Good point. We have now added in the following paragraph at the end of the background for the paper – which should make this point clearer.

GRIPP 2-SF is a short checklist for studies where PPI is a secondary or tertiary focus such as in a RCT, and GRIPP 2-LF is a longer checklist for studies where PPI forms the primary focus of a study, such as a paper primarily reporting the impact of PPI on the study. For GRIPP2-LF the entire paper

can be shaped by the guidance, with researchers selecting the items of relevance. With GRIPP2-SF researchers could present all the information in a short section or in a separate box.

R: While the article acknowledges the lack of consistent MESH terms, there is no acknowledgement of the way in which different ideologies / discourses or traditions impact on PPI. For example we might expect to see Ives et al cited as well as more recent publications that deal with critical aspects of PPI.

Ives, Jonathan, Sarah Damery, and Sabi Redwod. "PPI, paradoxes and Plato: who's sailing the ship?." *Journal of medical ethics* (2012): medethics-2011.

Similarly, Forbat, Liz, Gill Hubbard, and Nora Kearney. "Patient and public involvement: models and muddles." *Journal of clinical nursing* 18.18 (2009): 2547-2554.

Again, it may be that these conceptual issues are covered in the other article that has been submitted but I cannot tell.

A: The issues of conceptualisation and theoretical underpinnings of PPI are still poorly developed and we would expect greater debate and development of these areas as the PPI evidence base grows. Items in GRIPP2-LF ask authors to report on concepts and theory development around PPI. We have added a brief sentence in the conclusion with the references suggested alongside a report I published with UK-CRC which reviewed the evidence around conceptualisation and theoretical underpinnings of PPI:

With a growing evidence base, we expect greater discussion around the conceptualisation and theoretical underpinning of PPI to provide a greater insight into practices and processes (Ives 2013, Brett 2010, Forbat 2009).

R: PPI has long been a contested area and the impulse towards consensus is understandable but readers might learn more than is presented here currently from the challenges faced in resolving difference. This is glossed over in the description of the consensus face to face meeting but perhaps it is discussed in the main article. Materials were sent in advance of the consensus meeting but the authors should specify which the 'one or two key papers' were and reflect on whether and if so, how, these papers were thought to have influenced the discussion.

A: We agree that there are challenges faced in resolving the differences in the field of PPI. However, as the evidence base expands through use of the published reporting guidelines it is hoped that this will bring about greater consistency and fewer differences.

The key papers sent in advance of the consensus meeting were the following:

1. Brett J, Staniszewska S, Mockford C, Sandra Herron-Marx S, Hughes J, Tysall C, Suleman R (2014). Mapping the Impact of Patient and Public Involvement (PPI) on Health and Social Care Research: A systematic review. *Health Expectations*. 17(5) pp. 637–650
2. Mockford C, Staniszewska S, Griffiths F, and Herron-Marx S (2011). The impact of patient and public involvement (PPI) on UK NHS healthcare: a systematic review. *International Journal for Quality in Health Care*, (1353-4505)
3. Staniszewska S, Brett J, Mockford C, Barber R (2011). The GRIPP checklist: Strengthening the quality of patient and public involvement reporting in research. *International Journal of Health Technology Assessment in Health Care*, 27 (4): 391-399

R: Could the authors explain the need for GRIPP 2 and how frequently this might need to be updated?

A: The following sentence has been added to the end of the background information to explain the need for GRIPP2:

While the original GRIPP checklist represented an important starting point in creating high quality PPI reporting, its development drew on systematic review evidence, without broader input from the international PPI research community. Achieving consensus is now acknowledged as a crucial step in producing a reporting guideline [6]. GRIPP2 addresses this gap by developing consensus within the international PPI community.

It is unsure at this stage how often the guideline would have to be updated as it will depend on the speed of new knowledge in this area. We will update the searches every two years.

R: A weblink to HTAi Citizen and Patient Involvement Group would be helpful – perhaps as a footnote

A: A weblink has been added as a footnote to the relevant page

<http://www.htai.org/interest-groups/patient-and-citizen-involvement.html>

R: In the Delphi survey sub section of Methods, Round 3 changed concepts. It would be interesting to explain and discuss these concepts.

A: The changed concepts are reported in the GRIPP2 results paper, to be published in the BMJ.

R: Finally, as a researcher with an interest in public policy, I would simply urge caution in assuming that policy is necessarily based solely on 'rational decision making' (p.7), value judgements remain important.

A: We completely agree with this comment

## Reviewer 2

The reviewer's comments are in black, and our response to the reviewer's comments can be seen in red.

R: This is a methods manuscript describing the (further) development of a guideline for how papers should report patient and public involvement (PPI) in research.

Overall impression:

This is a well-written manuscript on an important topic. Since PPI in research projects is increasingly advocated for and sought after, the need for high quality reporting guidelines is evident. This manuscript seeks to answer to this need. While this manuscript has many qualities, I think that a revision would improve important aspects of the manuscript. My comments are as follows:

A: Thank-you

Major issues:

Patient and public involvement in research

R: 1. Throughout the manuscript it is not specified that the topic is PPI in research and not in general (as in health services etc.). The title is confusing since it states that the manuscript is about guidelines for reporting PPI, and does not specify that the subject is PPI in research in particular. I would recommend that the title is revised to specify this.

The sentence "Patient and Public Involvement in health and social care research" could also be interpreted to include all research on PPI, not only when patients or representatives from the public take part in research processes. This makes the Lay summary and the Abstract somewhat unclear, and this lack of specification is present throughout the manuscript.

A: Good point. We agree that it should be PPI in research, and we have changed the title to reflect this:

Reaching Consensus on Reporting Patient and Public Involvement (PPI) in Research: Methods and lessons learned from the development of reporting guidelines

We have also changed the first sentence of the lay summary to make it clearer that PPI in research is about working with researchers to prioritise, plan, design and conduct the research studies. The first sentence of the lay summary now reads:

Patient and public involvement (PPI) in health and social care research has grown over the past decade, with patients and the public working with researchers to help prioritise, plan, design and conduct research studies

Background:

R: 2. The description of the development process is well-written and in general easy to follow. It is clear how the systematic literature review, the Delphi process and the consensus meeting were conducted. However, I miss a description of the transition from the literature review to the Delphi process. The description "jumps" from the review directly to the GRIPP2 guideline items and the first stage of the Delphi process. How were the results from the updated literature review developed into the items in the first Delphi survey, and how did the researchers further develop the responses in the first survey into the second etc.? These methodological steps are important and are now lacking. (In the present manuscript, it is only described that "Two researchers analyzed the results of round 1 and free text comments were examined to inform any potential additional items." (Page 12, paragraph 2))

A: The details of the transition from the literature to the first GRIPP checklist is described in a previous paper (Staniszewska et al, 2011). The last paragraph about the systematic review in the methods sections reports summary detail of how the items for the Delphi were selected from the systematic review evidence. We have also referenced the first GRIPP paper (Staniszewska 2011) for readers to obtain further details of this transition.

We have now added a sentence at the end of the Delphi 1 sections to report the development of the responses in the first survey into the second:

Consensus was defined by the consistency of median scores (median  $\geq 8$  = high importance, median 6 or 7 = moderate importance, and median  $\leq 5$  = low importance), and the absence of significant issues noted in the text comments. Items reaching high importance or moderate importance were



selected for round 2 of the Delphi. Free text comments were analysed thematically to identify additional items for round 2.

The subsequent paragraphs describing the Delphi process have been adapted accordingly.

Discussion and conclusion:

R: 3. The first and second paragraph in this section is only repetition of the argument for doing the study, and should be omitted, except for the last two sentences, which sum up the aim of the paper.

A: This is a good point, and we have deleted all but the last two sentences from the start of the discussion. The first sentence has been integrated into the background section.

R: 4. Too much of the description in the Methods section is repeated in the part “Important aspects and lessons learned from the development process”. The repetition in this section should be removed and only the parts concerning aspects of lessons learned should be kept. This part of the manuscript should in general be shortened to avoid unnecessary repetition.

A: Good point. We have now edited the text to remove any repetition between the sections

R: 5. On page 20, last paragraph, results from the pilot study are referred to. The pilot study is described at page 11, second paragraph, but it is not described how the results from the pilot were used in the Delphi survey. I suggest that the information about the pilot study in the Discussion section is moved to the Methods section, since it is barely descriptions and not an actual discussion.

A: We agree that the pilot description sits better in the methods section. We have now moved the paragraph describing the outcomes of the pilot study into the methods section.

R: 6. From reading the list of points in “Strengths and limitations of this study” I anticipated more discussion of the role of the facilitator of the consensus meeting. This part of the discussion is short and very general, and includes no information on this specific meeting for this specific project. An addition here could increase the usefulness of this manuscript.

A: We have now expanded this paragraph about the important role of the facilitator, and added in a specific example:

Furthermore, a critical role in the success of the consensus group was that of the facilitator. Key aspects of this role were ensuring that participants of the group understand their roles, and adopting the listening stance so participants all felt listened to. This important role helped to ensure that balanced views were recorded through individual and group work. Facilitated groups develop greater consensus than user-driven groups [52,53]. However, highly structured facilitation can have an adverse effect on the consensus process, and element of flexibility in the process is recommended [52,53] A highly skilled facilitator was used to mediate the group process, and to ensure key and timely contributions from all members. The facilitator presented items where consensus decisions were needed, guided the participants to reach agreement, ensuring all have an opportunity to participate. For the items related to economic assessment and testing conceptual or theoretical models, where decisions on whether to include or not were difficult, the facilitator used problem solving techniques to finalise the decision. This involved the group drawing up a list of the pro's and con's of the item, and asking them to review and evaluate the list and then to re-evaluate their initial decision.

Minor issues:

R: 1. INVOLVE is introduced in the Background section without any references. This should be added.

A: We realise that INVOLVE should be acknowledged in the background information, and have put a footnote on this page to refer readers to the INVOLVE website. We have also referenced INVOLVE in the methods for Systematic Reviews section.

R: 2. On page 17, last paragraph is says "Evidence shows only an 8 % improvement in identification of relevant papers when all abstracts are reviewed by two researchers." It is unclear what the reference for this statement is. This should be clarified.

A: Good point – we have now put the reference after this sentence.

#### **VERSION 2 – REVIEW**

<b>REVIEWER</b>	Pam Carter University of Leicester, UK
<b>REVIEW RETURNED</b>	31-Jul-2017
<b>GENERAL COMMENTS</b>	The authors have now addressed comments outlined in my initial review.